Video-assisted thoracic surgery for pulmonary sequestration compared with posterolateral thoracotomy

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Objectives: Pulmonary sequestration is a rare congenital malformation of the lungs. This study aims to evaluate the effectiveness of video-assisted thoracic surgery for the treatment of pulmonary sequestration in a larger series compared with posterolateral thoracotomy.

Methods: The files of 42 patients with pulmonary sequestration treated via video-assisted thoracic surgery (18 cases) and posterolateral thoracotomy (24 cases) between September 2005 and May 2012 from a single institute were retrospectively reviewed. Data were collected regarding the patient demographics, medical history, preoperative investigations, intraoperative findings, and postoperative course.

Results: All sequestration lung lesions were found in the lower lobes (31 on the left, 11 on the right), with feeding arteries arising from the thoracic aorta (34 cases) and the abdominal aorta (8 cases). Thirty-nine cases of sequestration were intralobar, and only 3 cases were extralobar. All patients achieved successful resection (including 37 lobectomies, 2 pneumonectomies, and 3 resections of the extralobar lesion). In the video-assisted thoracic surgery group, 1 case was converted to thoracotomy because of an injury to the aberrant artery; 1 case had injury to the left lower pulmonary vein and 1 case had injury to the aberrant artery, which were successfully treated without conversion. No significant differences were found between the 2 groups (video-assisted thoracic surgery vs posterolateral thoracotomy) in terms of the duration of operation, blood loss, amount of chest drainage, duration of chest drainage, length of postoperative hospital stay, and complications.

Conclusions: Video-assisted thoracic surgery resection for pulmonary sequestration is feasible, although it should be performed by an experienced surgeon with awareness of the potential risk of severe vascular injury. (J Thorac Cardiovasc Surg 2013;146:557-61)

Pulmonary sequestration (PS) is a rare congenital malformation characterized by nonfunctional lung tissue separated from the normal tracheobronchial tree and fed by an aberrant systemic artery.[1] PS accounts for 0.15% to 6.45% of all pulmonary malformations.[2] The following types of PS have been recognized: intralobar pulmonary sequestration (ILS), which is an abnormal region within the normal pulmonary parenchyma without its own pleural covering, and extralobar pulmonary sequestration (ELS), which has its own pleural covering. PS is supplied by an aberrant systemic artery, most frequently from the descending thoracic or abdominal aorta. Venous drainage is usually toward the pulmonary veins for ILS and toward the systemic venous system for ELS.[3] Despite its being a benign-condition, the potential complications of PS are serious and may include recurrent pulmonary infection, hemoptysis, congestive heart failure, and tumorigenesis.[4,5] Therefore, the main form of treatment has always been surgical excision even for asymptomatic patients with PS.

The conventional surgical approach for the resection of PS is through a posterolateral thoracotomy (PLT) approach. As an alternative, video-assisted thoracic surgery (VATS) has been increasingly recognized as an equally effective, minimally invasive approach for major lung resection; however, the VATS approach for PS treatment has only been reported in single case reports or in small case series as the definitive treatment.[6-8] We report one of the largest case series of PS, which consists of 42 patients (including 18 patients receiving the VATS approach) from a single hospital, to evaluate the presentation and outcomes of PS. The effectiveness of the VATS approach was likewise evaluated with respect to its feasibility, safety, and complications.

MATERIALS AND METHODS

We retrospectively reviewed the files of 18 patients with PS treated via the VATS approach in the Thoracic Department of the West China Hospital, Sichuan University, between September 2005 and May 2012. To better evaluate the effectiveness of the VATS approach, we reviewed the files of 24 patients who received successful resection for PS via the PLT approach during the same period at the same hospital. Data from each patient were collected regarding the patient demographics, medical history, preoperative investigations, intraoperative findings, and postoperative course.

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Abbreviations and Acronyms

CT = computed tomography
ELS = extralobar pulmonary sequestration
ILS = intralobar pulmonary sequestration
PLT = posterolateral thoracotomy
PS = pulmonary sequestration
VATS = video-assisted thoracic surgery

The protocol of the study was approved by the institutional review board of West China Hospital. Informed consent of the operation was obtained from all patients before surgery.

All patients were initially selected on the basis of their medical history and presumptive chest computed tomography (CT) manifestation. During the earlier stages of this study, more surgical resections were performed following the PLT approach. With the increased experience with the VATS procedure, our criteria for the selection of patients who received the VATS approach became more similar to those for the PLT approach. Thus, more patients received VATS in the later stages of this study. All procedures were conducted under single lung ventilation in a lateral decubitus position. First, the feeding artery from the aorta was meticulously dissected from the surrounding tissue around or within the inferior pulmonary ligament and cut using a stapling device. Specifically, if the aberrant artery was of a large caliber or seemed to be fragile, we proximally ligated the artery with silk suture before cutting it with a stapling device to ensure a solid stump. We inclined to anatomize the aberrant artery closer to the side of the lung tissue. Subsequently, anatomic lobectomy or pneumonectomy was performed for patients with ILS, and excision of the ELS was performed for patients with ELS. The diameter of each aberrant artery was measured immediately after the sequestrated lesion been retrieved. One chest tube was placed at the end of the procedure, but this tube was removed if no air leak was recognized and the daily output was less than 200 mL. Patients with no main complications were discharged from the hospital.

Data were analyzed using SPSS version 16.0 (SPSS Inc, Chicago, Ill). Categoric variables were analyzed using Pearson's chi-square test or Fisher exact test. Continuous variables were expressed as the mean ± standard deviation and analyzed using the Mann–Whitney Wilcoxon test.

RESULTS

The mean age at surgery was 35.9 years (range, 15-61 years). A comparison of the baseline demographic characteristics and perioperative clinical findings between the VATS and PLT groups is shown in Table 1. There were more female patients in the VATS group ($P < .05$), whereas there were no other significant differences between the 2 groups in terms of baseline demographic characteristics. Among the 42 patients, 34 (80.95%) presented signs and symptoms of pulmonary disease: recurrent pneumonia (cough, expectoration/purulent sputum, or fever; VATS group: 8 cases; PLT group: 8 cases), hemoptysis (VATS group: 2 cases; PLT group: 5 cases), thoracic pain (VATS group: 1 case; PLT group: 6 cases), respiratory distress (VATS group: 1 case; PLT group: 1 case), and pneumothorax episodes (VATS group: 1 case; PLT group: 1 case). Eight patients (19.05%) did not present any symptoms. Preoperative CT scans revealed signs of mass lesions (23 cases), cystic lesions (14 cases), and pneumonic lesions accompanied with bronchiectasis (5 cases). With multislice CT scanning and 3-dimensional reconstruction technique, 30 patients (71.43%) were diagnosed preoperatively with an obvious aberrant artery arising from the aorta (Figure 1). Other patients were misdiagnosed with pulmonary cysts, lung cancer, and other kinds of infectious disease. Seven cases of comorbidity were identified, namely, 3 cases in the VATS group (1 case each of pneumothorax, achalasia, and chronic hepatitis B) and 4 cases in the PLT group (1 case of pneumothorax, 1 case of esophageal diverticulum, and 2 cases of hypertension). All diagnoses were confirmed by intraoperative findings and postoperative histologic examination. All patients with ILS had intraoperative and postoperative antibiotic prophylaxis.

No significant differences were found between the 2 groups in terms of intraoperative findings. Patients with PS were generally found to have varying degrees of pleural adhesions. All sequestrated lung lesions were found in the right or left lower lobes: 31 cases in the left lower lobe and 11 cases in the right lower lobe. Among the 42 cases of sequestration, 39 were intralobar and only 3 were extralobar. The feeding artery varied from 0.3 to 2 cm in diameter, with a mean diameter of (0.72 ± 0.34) cm. The main vascular supply to the lesions arose from the thoracic aorta in 80.9% of the cases (34/42) and from the abdominal aorta in 19.1% of the cases (8/42). Two patients in the VATS group were confirmed to have 3 supplying arterial branches. Venous drainage of all 39 ILS cases was through the pulmonary vein, whereas drainage of the 3 ELS cases was via the azygos vein on the right and hemiazygos vein on the left.

In the VATS group, 15 patients underwent anatomic lobectomy. One case had excision of the ELS lesion and bullae resection in the left upper lobe because of recurrent pneumothorax. One case had anatomic lobectomy and Heller's myotomy for achalasia. One case underwent excision of the ELS lesion with conversion to thoracotomy caused by injury of the aberrant artery hidden in the scarred tissue, with blood loss of approximately 1200 mL. In the PLT group, 21 patients underwent anatomic lobectomy. Two cases had a pneumonectomy due to pulmonary hypoplasia with widespread cystic degeneration, 1 of which with additional excision of the esophageal diverticulum. One case underwent excision of the ELS lesion. Aside from 1 case with conversion, 2 more injuries occurred in the VATS group. One case had injury to the left lower pulmonary vein that was successfully treated with the suction-compressing angiorrhaphy technique[9] without conversion to thoracotomy and with blood loss of approximately 300 mL. The other injury was to an aberrant artery that was buried in the inferior pulmonary ligament and had not been presumptively identified by the preoperative CT scan. The aberrant artery was accidentally divided by hook cauterization. Two long forceps were immediately used to control the proximal and distal ends. The stapling device and titanium

TABLE 1. Preoperative characteristics of patients and perioperative clinical data

	VATS (n = 18)	PLT (n = 24)	*P* value
Sex (F:M)	12:6	6:18	.007
Age (y)	35.8 ± 14.2	35.9 ± 10.3	NS
Presence of preoperative symptoms (Yes:No)	13:5	21:3	NS
CT manifestation (Ma:Cys:Pneu)	9:6:3	14:8:2	NS
Comorbidity (Yes:No)	3:15	4:20	NS
Preoperative diagnosis (Yes:No)	14:4	16:8	NS
Site of sequestration (RLL:LLL)	4:14	7:17	NS
Type of sequestration (ELS:ILS)	2:16	1:23	NS
Aberrant artery origin (thoracic aorta:abdominal aorta)	15:3	19:5	NS
Venous drainage (pulmonary vein:systemic vein)	16:2	23:1	NS
Mean diameter of aberrant artery (cm)	0.64 ± 0.23	0.77 ± 0.40	NS
Duration of operation (min)	133.06 ± 42.26	142.50 ± 28.80	NS
Blood loss (mL)	186.11 ± 279.35	117.08 ± 60.18	NS
Chest drainage amount (mL)	391.67 ± 261.92	441.67 ± 194.30	NS
Duration of chest drainage (d)	2.72 ± 0.83	2.92 ± 0.78	NS
Postoperative hospital stay (d)	5.33 ± 1.75	5.83 ± 2.01	NS
Complications (Yes:No)	2:16	1:23	NS

VATS, Video-assisted thoracic surgery; *PLT*, posterolateral thoracotomy; *CT*, computed tomography; *Ma*, mass; *Cys*, cystic lesion; *Pneu*, pneumonic lesion; *RLL*, right lower lobe; *LLL*, left lower lobe; *ELS*, extralobar sequestration; *ILS*, intralobar pulmonary sequestration; *NS*, nonsignificant.

clips were then used to handle the proximal and distal ends, respectively. No short-term postoperative mortality was identified. Complications included 2 cases of pneumonia (1 each from the VATS and PLT groups) and 1 minor wound infection in the VATS group. Two cases in the PLT group were confirmed to have secondary infections by *Mycobacterium tuberculosis* and *Aspergillus* via postoperative pathologic examination, which were treated with antituberculosis and antifungal regimens, respectively. No case of bronchial or pulmonary malignant transformation of the sequestration was identified. The mean length of postoperative hospital stay was 5.62 ± 1.90 days (range, 3-13 days). As shown in Table 1, no significant differences were found between the 2 groups in terms of the duration of operation, blood loss, amount of chest drainage, duration of chest drainage, length of postoperative hospital stay, and occurrence of complications. Long-term postoperative follow-up was not analyzed. Among the patients who returned to the outpatient department, no recurrences or further complications occurred.

DISCUSSION

PS is a benign condition that can be completely asymptomatic or accompanied by complications. However, symptoms are not specific; it is hard to make the diagnosis preoperatively with only a history of disease. ILS should not be ignored if there is a recurrent infection in the lower lobes.[10] Nowadays, with novel radiologic modalities such as spiral CT and magnetic resonance angiography, most PS cases can be easily diagnosed preoperatively.[11,12] However, because CT angiography or 3-dimensional reconstruction technique was not routinely performed for every patient in the earlier stages of the study, there were

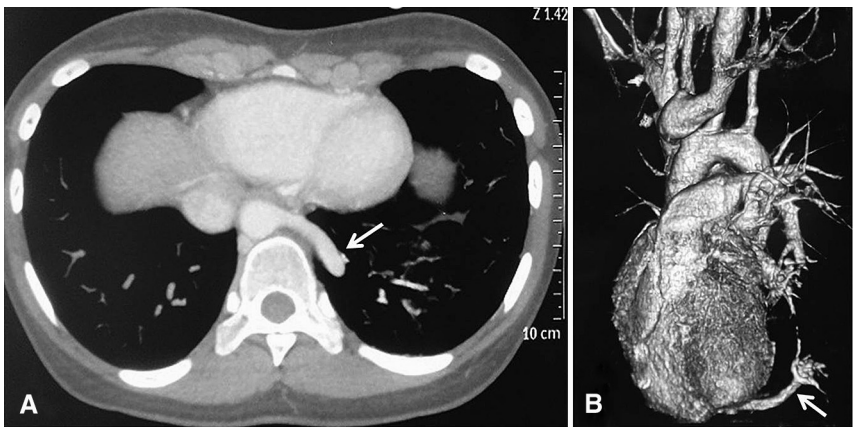

FIGURE 1. Aberrant artery (*white arrow*) originating from the thoracic aorta, as shown by CT scanning (A) and 3-dimensional reconstruction (B).

still 12 cases of preoperative misdiagnosis, which might result in a higher risk of unanticipated vascular injury.

Resection is generally accepted as the definitive treatment of choice for PS.[6,7,13,14] In the present series, we achieved complete resection of the sequestrated lesion for all patients. We performed a lobectomy for most ILS cases because of the size or location of the lesion. However, we are not averse to sublobar resection for PS, as reported by other experts,[7,15] if the patient is suitable for an adequate wedge resection or anatomic segmentectomy. Two cases with widespread cystic degeneration required pneumonectomy to remove the total nonfunctioning lung. Resection through PLT remains the most efficient established approach for definitive resection of PS. However, the feasibility of VATS resection for PS has been shown in single case reports or small case series since 1994.[6-8,16,17] To the best of our knowledge, this is the largest report of thoracoscopic treatment for PS compared with the conventional PLT approach.

The main difficulty during resection of a PS lesion via VATS lies in the identification of the aberrant artery. In this study, the aberrant artery was mostly present in the inferior pulmonary ligament. Inflammatory changes caused by recurrent infections were always present. Dense adhesions accompanied with more proliferative vessels would make the surgical field bloody and blurred during dissection. The interchangeable use of the electrocoagulation hook and ultrasonic dissector effectively kept the surgical field clean, thereby facilitating the identification of the aberrant artery. Preoperative imaging helped us anticipate the localization of the target vessel. After the aberrant artery had been identified and dissected from peripheral tissues, a stapling device was introduced to cut the artery (Figure 2). The aberrant artery would be thickened or fragile because of the recurrent infections. For these thickened or fragile vessels, we often proximally ligated the artery with silk suture before cutting it with a stapling device to ensure a solid stump. Three accidents occurred during the practice of VATS. All these accidents were due to the confusion in the local anatomy as caused by inflammatory changes, previous unawareness of the existence of the aberrant artery, and human error. These complications should be avoided in future studies, and the identification of the aberrant artery should be performed with utmost care at the beginning of the procedure, especially for the misdiagnosed patients. Moreover, if unanticipated injuries to vessels occur, a clear thoracoscopic view should be maintained. For injury to pulmonary vessels with lower blood pressure, bleeding can be appropriately handled using the novel method of angiorrhaphy described in our former study.[9] For injury to the aberrant artery, the proximal end should be clamped quickly in response to the excessive bleeding caused by high blood pressure. We successfully controlled 1 case of arterial bleeding because the proximal end was long enough for the introduced stapling device to cut it; thus, anatomization of the aberrant artery should be performed as close to the lung tissue as possible at the beginning to reserve a relative long proximal end for handling bleeding if unanticipated vascular injury happens. Of note, there will always be circuitous and thickened bronchial arteries, which will lead to intraoperative or postoperative bleeding if they are not handled appropriately. Antecedent management of these troublesome arteries would be meaningful for the following transection of the bronchus. Titanium clips or an ultrasonic dissector is always used to handle these arteries in our practice.

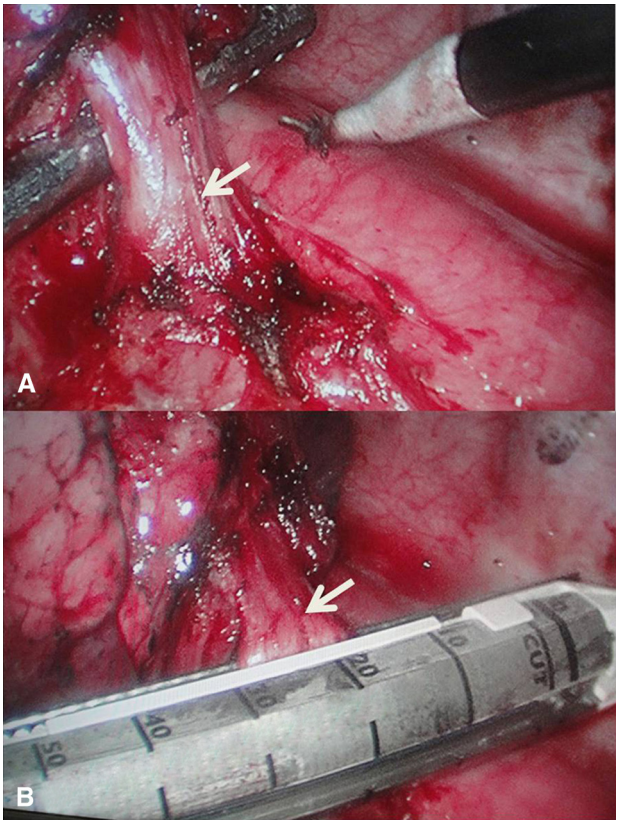

FIGURE 2. Thoracoscopic identification (A) and cutting (B) of the aberrant artery (*white arrow*).

Although more of the patients during the earlier stages of this study were treated via the PLT approach, we preferentially chose the VATS approach during its later stages. Therefore, selection bias must exist. Although we had no inclination to select VATS for female patients, there was still a significant difference of sex between the 2 groups. Although the results suggested that VATS resection for PS was feasible, there is still concern for potentially life-threatening vascular injury, as occurred in this study. Except for meticulous operation, preoperative awareness of the existence of the aberrant artery was the key point to reduce the risk of intraoperative accidents. In the future, we would like to improve our preoperative workup to avoid misdiagnosis.

In addition, the VATS procedure for PS should be performed by experienced surgeons, and the application of VATS should not be pursued as something trendy.

CONCLUSIONS

This study demonstrated that the VATS resection for PS was feasible. However, the VATS procedure should be encouraged only for experienced surgeons because of the potential risk of life-threatening vascular injury. Lobectomy remains the main option, whereas sublobar resection should be considered only when absolutely necessary.

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